A Case of Aneurysm on a Persistent Hypoglossal Artery Treated by Endovascular Coiling

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Summary

We describe a 22-year-old woman admitted to hospital in emergency with nuchal headache and vomiting. CT scan disclosed subarachnoid hemorrhage. Digital subtraction angiography with three-dimensional rotational acquisitions showed a ruptured aneurysm of a right persistent primitive hypoglossal artery as the cause of symptoms and hemorrhage. The patient was successfully treated with endovascular coiling of the aneurysm. This is the second literature report describing endovascular treatment in this unusual condition.

Introduction

The persistent hypoglossal artery is the second most common carotid-vertebrobasilar anomalous anastomosis (the first being the trigeminal artery) [1]. It originates from the extracranial internal carotid artery as a large collateral branch, ascends vertically, passes through the hypoglossal canal and continues as the basilar trunk. Hypoplasia of vertebral arteries is frequently associated, so that the persistent hypoglossal artery is often the only functional vessel supplying blood to the posterior circulation. The anomaly is often asymptomatic, but it may cause paralysis of the XII cranial nerve and IX nerve neuralgia. Definitive diagnosis can be established by CT or MR angiography when an anomalous vessel is visualized in an enlarged hypoglossal canal [2].

Case Report

A 22-year-old woman was admitted to our department with sudden nuchal headache and vomiting. She had been transferred in emergency from a nearby hospital where she had undergone a CT brain scan that disclosed subarachnoid and intraventricular hemorrhage. No other neurological signs were present and her history was unremarkable. Hunt and Hess scale of subarachnoid hemorrhage was 3.

Angiography was performed in emergency and revealed a persistent hypoglossal artery originating from the internal carotid artery at the level of C2, ascending to the base of the skull with a slight posterior bend, entering the skull via the hypoglossal canal and continuing on the midline as the basilar trunk. An aneurysm was evident on the intracranial portion of the hypoglossal artery. Rotational acquisitions allowed three-dimensional reconstructions and an optimal visualization of the aneurysm neck and measurement of the sac (Figure 1). The widest diameters of the aneurysm measured 8.5 mm and 6.8 mm; the neck was relatively tight, measuring 3.3 mm. Multiplanar axial reconstructions yielded an accurate depiction of the vessel's anatomical features and its course in the hypoglossal canal (Figure 2).

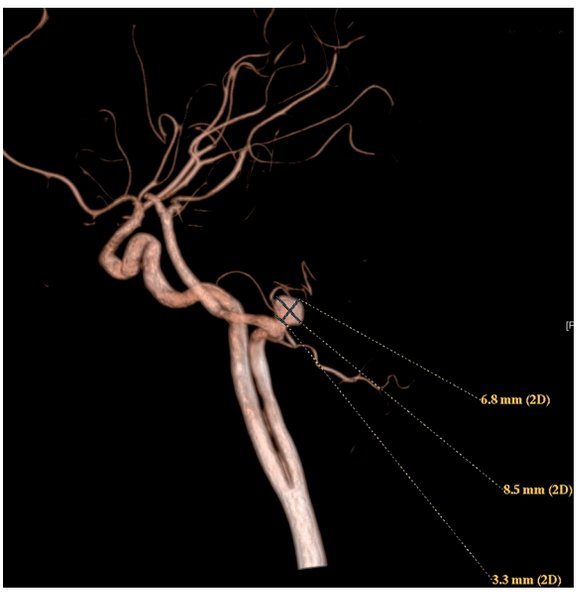

Figure 1 Three-dimensional reconstructions after rotational acquisition allow a good depiction of the aneurysm and measurement of its diameters.

Both vertebral arteries were present but hypoplastic. On the left, the posterior inferior cerebellar artery originated from the ipsilateral vertebral trunk and the anterior inferior cerebellar artery from the basilar trunk. On the right, a dominant posterior inferior cerebellar artery originated from the hypoglossal artery. The two posterior communicating arteries were not visible.

Endovascular Treatment

The embolization procedure was carried under general anesthesia, via a 6F sheath in the right femoral artery. A 6F Guider (Boston Scientific, Natick, MA, USA) guide catheter was located in the hypoglossal artery and then an Excelsior SL-10 90° angled microcatheter (Boston Scientific, Natick, MA, USA) was coaxially placed over a 0.012-inch microguide wire (GT Terumo, Tokyo, Japan) into the aneurysm sac. The patient was heparinised with 3000 IU i.v., and six coils (two GDC and four Matrix, Boston Scientific, Natick, MA, USA) were delivered into the aneurysm. Final controls revealed a complete exclusion of the aneurysm from the circulation and a good packing density inside the sac (Figure 3), with patency of all the vessels of the posterior circulation. One year after treatment, the patient is in good health and MR angiography follow-ups have shown no signs of recanalization of the aneurysm.

Discussion

In the early stages of embryonic development the hindbrain circulation is supplied by four pairs of arterial anastomoses connecting the dorsal aorta (future internal carotid) with the longitudinal neuronal arteries located dorsally on the surface of the hindbrain. These anastomoses (named trigeminal, otic, hypo-

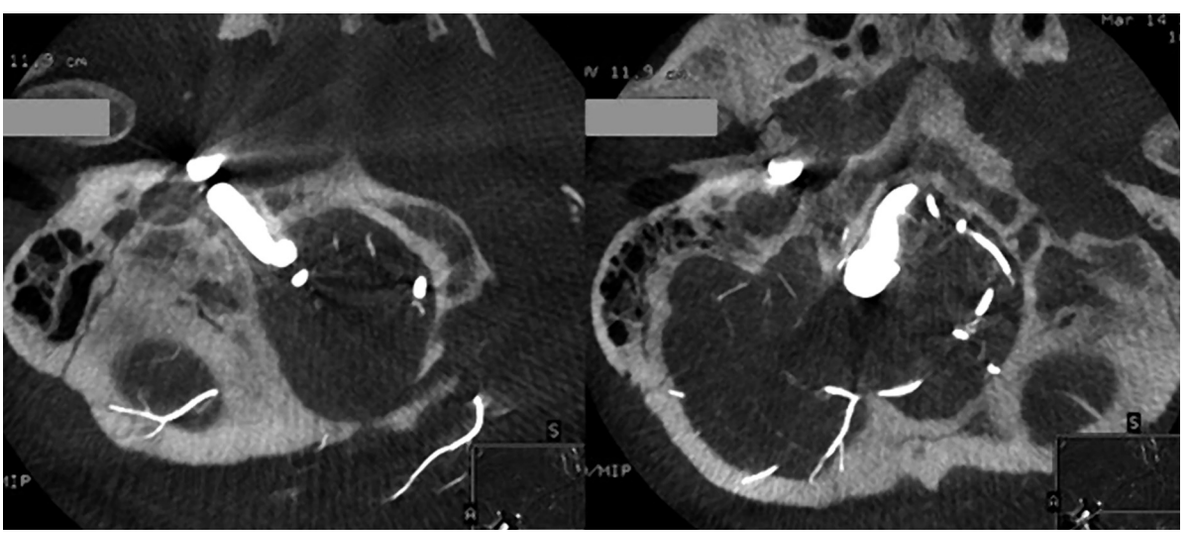

Figure 2 Multiplanar axial reconstructions after rotational angiography permit an optimal visualization of the anomalous vessel at the skull base, the aneurysm and the other arteries of the posterior circulation.

glossal and proatlantal intersegmental arteries) form when the embryo measures about 4-5 mm (approximately at the fifth week of gestational age) and persist for about one week. They start to regress when the posterior communicating arteries form (at the 5-6 mm embryo stage), and at the same time the two longitudinal neural arteries partially merge to form the basilar artery. At the 7-12 mm embryo stage (sixth week of gestational age) the vertebral arteries form and supply the caudal part of the longitudinal neural arteries. At this stage the final vertebrobasilar system is formed and the anastomoses with the carotid system have normally regressed [3].

Holmin et Al suggested that these embryonic vessels (and specifically the hypoglossal artery) could be identified as part of segmental entities, helping to explain peculiar pathologies affecting both anterior and posterior brain circulations in post-natal life [4]. These anastomoses can persist in the adult life, the most common being the persistent trigeminal artery, while the rarest is the otic artery whose existence remains a matter of controversy [5].

A persistent primitive hypoglossal artery has been reported to occur with a frequency of 0.1% at MR angiography in a study of 900 patients [2]. The artery arises from the origin of the cervical internal carotid artery, joins the 12th cranial nerve and enters the posterior cranial fossa via the hypoglossal canal. It can join the contralateral vertebral artery or, at a higher position, the basilar artery [5]. The latter occurrence was observed in our patient, with the hypoglossal artery taking over both ipsilateral PICA and AICA territories.

The anomaly can be asymptomatic or give rise to cranial nerve paralysis or neuralgia [6]. Some claim that it can be responsible for hemodynamic changes leading to vertebrobasilar insufficiency, cerebral infarction or intracranial aneurysms formation [7]. An association between persistent hypoglossal artery and intracranial aneurysms has been reported [8-11]. As suggested by Mazighi et Al, vascular anomalies may not be directly responsible for aneurysm formation, but could be a marker of an abnormal development of congenital origin. Hence, the vascular anomaly corresponds to a segmental vulnerability which can predispose to aneurysm formation and growth [12].

The presence of an aneurysm on the hypoglossal artery itself is a very rare condition in the general population [13-17], whereas it occurs with a frequency of about 26% when a persistent hypoglossal artery is present [13]. The relation between the anomaly and the aneurysm may be explained by the persistence of immature endothelial cells in the intima of the embryonic artery and congenitally impaired apoptosis. A congenital structural defect in the vessel wall may also account for the tendency of these lesions to rupture in the subarachnoid space [18], as in our patient and the majority of reported cases.

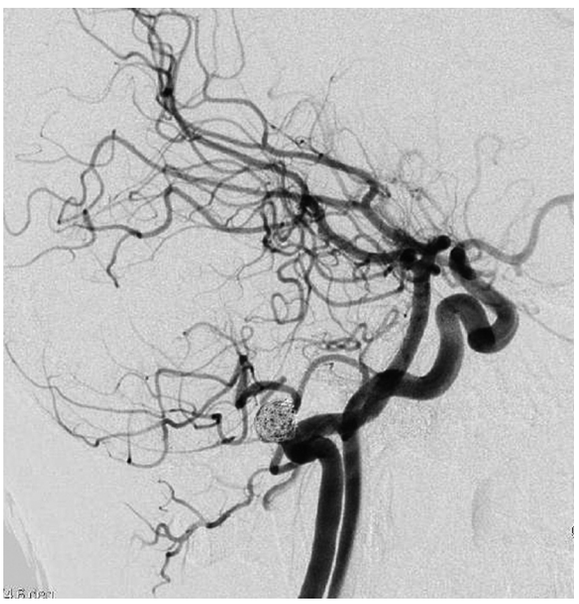

Figure 3 Post-treatment control shows a good packing density inside the lesion and a complete exclusion from the bloodstream.

Diagnosis and treatment of persistent hypoglossal artery are extremely important since the hypoglossal artery is often the only vessel supplying blood to the basilar trunk and posterior circulation in these patients. This was not the case of our patient in whom both vertebral arteries were present.

Literature reports of persistent hypoglossal artery were recently reviewed by Baltsavias et Al with a description of associated lesions and treatment procedures.

Baltsavias et Al were also the first to describe an endovascular treatment in a hypoglossal artery aneurysm [19]. To our knowledge, our patient is the second case in whom endovascular therapy with coiling was carried out to treat a ruptured aneurysm of the hypoglossal artery. A recent case report by Baldi et Al de-

scribed the stent-assisted coiling of an unruptured wide-necked aneurysm located on a persistent primitive hypoglossal artery in a patient with pulsatile tinnitus [20].

Accurate preoperative demonstration of the lesion (location, diameters, neck, morphology) is mandatory to perform a safe treatment, and is obtained by CT angiography with three-dimensional reconstructions [20] or 3D rotational angiography. In our patient, three-dimensional rotational angiography provided a complete assessment of the lesion and the possibilities of treatment.

Conclusions

Our endovascular procedure for coil embolization of a persistent hypoglossal artery aneurysm was safe, uneventful and effective with a good result and an excellent outcome at one year. Preoperative three-dimensional rotational angiography provided an optimal depiction of the diameter and global morphology of the lesion. Endovascular coiling appears to be a valid alternative to surgery for hypoglossal aneurysms and awaits further confirmation in future reports.

References

1 Caldemeyer KS, Carrico JB, Mathews VP: The radiology and embryology of anomalous arteries of the head and neck. Am J Radiol 170: 197-203, 1998.
2 Uchino A, Sawada A et Al: MR angiography of the anomalous branches of the internal carotid artery. Am J Radiol 181: 1409-1414, 2003.
3 Padget DH: Designation of the embryonic intersegmental arteries in reference to the vertebral artery and subclavian stem. Anat Rec 119: 349-356, 1954.
4 Holmin S, Ozanne A et Al: Association of cervical internal carotid artery aneurysm with ipsilateral vertebrobasilar aneurysm in two children: a segmental entity? Childs Nerv Syst 23: 791-798, 2007.
5 Lasjaunias P, Berenstein A, ter Brugge KG: Craniocervical junction. In: Lasjaunias P, Berenstein A, ter Brugge KG: Surgical neuroangiography. Volume 1: Clinical vascular anatomy and variations - second edition. Springer-Verlag, Berlin 2001: 165-259.
6 Yilmaz E, Ilgit E, Taner D: Primitive persistent carotid-basilar and carotid-vertebral anastomoses: a report of seven cases and a review of the literature. Clin Anat 8: 36-43, 1995.
7 De Caro R, Parenti A, Munari PF: The persistent primitive hypoglossal artery: a rare anatomic variation with frequent clinical implications. Ann Anat 177: 193-198, 1995.
8 Pasco A, Papon X et Al: Persistent carotid-vertebrobasilar anastomoses. How and why differentiating them? J Neuroradiol 31: 391-396, 2004.
9 Kanematsu M, Sato K et Al: Ruptured aneurysm arising from a basilar artery fenestration and associated with a persistent primitive hypoglossal artery. Case report and review of the literature. J Neurosurg 101: 532-535, 2004.
10 Huynh-Le P, Matsushima T et Al: Persistent primitive hypoglossal artery associated with proximal posterior inferior cerebellar artery aneurysm. Surg Neurol 62: 546-551, 2004.
11 Hatayama T, Yamane K et Al: Persistent primitive hypoglossal artery associated with cerebral aneurysm and cervical internal carotid artery stenosis - Case report. Neurol Med Chir (Tokio) 39: 372-375, 1999.
12 Mazighi M, Porter PJ et Al: Vascular anomalies and the risk of multiple aneurysms development and bleeding. Interventional Neuroradiology 8: 15-20, 2002.
13 Yamamoto S, Sunada I et Al: Persistent primitive hypoglossal artery aneurysms - report of two cases. Neurol Med Chir (Tokio) 31: 199-202, 1991.
14 Al-memar A, Trush D: Unilateral hypoglossal nerve palsy due to aneurysm of the stump of persistent hypoglossal artery. J Neurol Neurosurg Psychiatry 64: 405,1998.
15 Duffill J, Lang DA, Dwyer GN: Subarachnoid haemorrhage in a child from an aneurysm of a persistent primitive hypoglossal artery. Br J Neurosurg 10: 607-610, 1996.
16 Kanai H, Nagai H et Al: A large aneurysm of the persistent primitive hypoglossal artery. Neurosurgery 30: 794-797, 1992.
17 Murayama Y, Fujimoto N, Matsumoto K: Bilateral persistent primitive hypoglossal arteries associated with a large ruptured aneurysm on one side. Surg Neurol 24: 498-502, 1985.
18 Krings T, Lasjaunias PL et Al: The aneurysmal wall. The key to a subclassification of intracranial arterial aneurysm vasculopathies? Interventional Neuroradiology 14: 39-47, 2008.
19 Baltsavias GM, Chourmouzi D et Al: Ruptured aneurysm of a persistent primitive hypoglossal artery treated by endovascular approach - case report and literature review. Surg Neurol 68: 338-343, 2007.
20 Baldi S, Zander T et Al: Stent-assisted coil embolization of a wide-neck aneurysm of a persistent primitive hypoglossal artery. Cardiovasc Intervent Radiol DOI 10.1007/s00270-008-9415-4.

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